Enhancing Patient-Centred Care through Knowledge Co-creation and Empowerment

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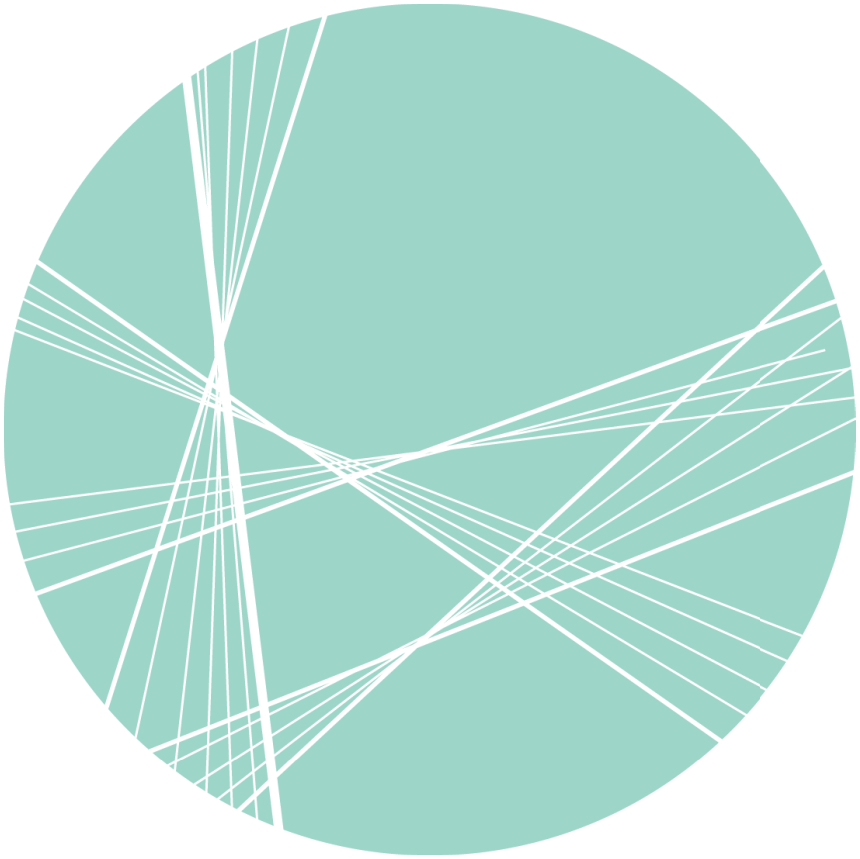

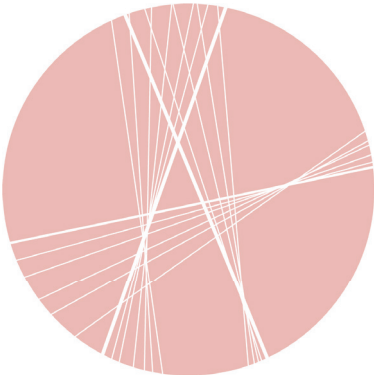

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EMPOWERING PATIENTS AND HEALTH PROFESSIONALS TO ADDRESS SEXUAL HEALTH

ABSTRACT[15]

Next to chronic incontinence and constipation, patients with Anorectal Malformations and Hirschsprung Disease regularly cope with sexual problems. Previous research shows that sexual health support for these patient groups is, however, lacking. Despite the development of various tools for health professionals to stimulate communication about sexual health, sexuality is still an underestimated subject in current care services. Therefore this study aims to develop a tool that is aligned with patients' and health professionals' needs to address sexual health in the context of Anorectal Malformations and Hirschsprung disease. To develop the tool, a multi-phased participatory action research was conducted in the Netherlands. First, an inventory of needs was made through interviews (n=22), three online focus groups (n=24) and a questionnaire (n=38). Patient and professional empowerment models were used to analyse and structure the data. Subsequently, four co-creation sessions (n=13) were organised to translate the needs into an empowerment tool. The functionality of the tool was assessed via a questionnaire (n=34). Patients indicated a need to feel not like a patient, receive an empathetic response and obtain knowledge about the (possible) sexual problems and possibilities for self-management and professional support. Health professionals were often not aware of the importance of sexual health and how sexologists could be deployed and were insecure about who is responsible for delivering sexual health. In addition, they missed the knowledge about the (possible) sexual problems and possible solutions, conversational skills and time to address sexual health. An empowerment tool (in the form of a website) was developed for patients, their parents and health professionals to stimulate awareness, fill current knowledge gaps and show possibilities for support. By simultaneously educating these groups, the website is expected to change restrictive attitudes towards sexual health and help the legitimization of the topic needed for the allocation of resources and sexologists' involvement in current care pathways.

[15] Petit-Steeghs, V., Mogami-Asselin, G.I.K., Nijenkamp, M.D., Spoel, M., Broerse, J.E.W., Pittens, C.A.C.M. on behalf of Consortium 'Support Psychosexual problems Congenital Colorectal malformations'(SPCC) consisting of VU University, Erasmus MC Rotterdam, Radboud UMC Nijmegen, UMC Utrecht, Amsterdam UMC, the patient organisations of Anorectal Malformations and Hirschsprung disease and the Dutch genetic alliance VSOP. Empowering patients and health professionals to address sexual health in the context of Anorectal Malformations and Hirschsprung disease. *Journal of Qualitative Health Research*
The original abstract of the manuscript has been adapted for this thesis.

8.1 | INTRODUCTION

Globally, the congenital diseases Anorectal Malformations (ARM) and Hirschsprung disease (HD) occur one in every 5,000 live births (Levitt and Peña, 2007). These diseases are respectively caused by the incomplete or insufficient development of the anus or rectum (Holschneider and Puri, 2008) or the absence of enteric ganglia along the intestine (Amiel *et al.*, 2008). Although ARM and HD are often diagnosed and treated during the neonatal period, patients often experience long-term gastrointestinal morbidity such as chronic incontinence and constipation (Teitelbaum and Coran, 2008; Stenström *et al.*, 2014). In addition, psychological problems are frequently observed, such as shame due to faecal incontinence and a negative body image as a result of wearing a stoma (Weerakoon, 2001; Manderson, 2005) or dilatations during childhood (Witvliet *et al.*, 2017; Athanasakos *et al.*, 2006). These psychological problems may make patients hesitant to seek friendships and sexual relationships in adulthood (Witvliet *et al.*, 2018), and cause sexual impairment such as erectile dysfunction, vaginism or dyspareunia (van den Hondel *et al.*, 2015; Schmidt *et al.*, 2012; Konuma *et al.*, 2006). Sexual impairment could also be the result of post-operative problems, such as scar tissue, or constrictions in the genital area due to urogenital anomalies [7-9]. These problems[16] can negatively affect the sexual health of these patient groups (van den Hondel *et al.*, 2015; Rigueros Springford *et al.*, 2016; Kyrklund *et al.*, 2016).

Good sexual health is essential for a person's physical and emotional well-being and thus quality of life (Grano *et al.*, 2012). The World Health Organization (2006: p.5) defines sexual health as "*a state of physical, emotional, mental and social well-being in relation to sexuality; it is not merely the absence of disease, dysfunction or infirmity*" (Organization, 2006). A study by van den Hondel *et al.* (2015) highlighted, however, that patients with ARM and HD experience a need for support in managing their sexual problems. Yet, there is a lack of attention to sexual health in clinical practice (Witvliet *et al.*, 2017; Schmidt *et al.*, 2012; Organization, 2006). van den Hondel *et al.* (2015) showed that 93.5% of the patients they questioned, did not receive any support – in the form of counselling or patient education – regarding (future) sexual problems. Of these patients 60% (ARM) and 59% (HRM) felt a need for education (van den Hondel *et al.*, 2015). In addition, Witvliet *et al.* (2017) found that only one out of 168 patients with ARM and HD was referred to a specialist for sexual-related issues. Various authors suggested the need for better follow-up during adulthood to improve support (Stenström *et al.*, 2014; Witvliet *et al.*, 2018). Previous studies concerning other diseases showed that neither patients nor health professionals initiate the discussion on sexual health (McInnes, 2003; Traumer *et al.*, 2019). Patients feel reluctant to ask for help regarding sexual issues (Hautamäki *et al.*, 2007; Vermeer *et al.*, 2015) due to a lack of trust (Fitch *et al.*, 2013) and feelings of embarrassment (Traumer *et al.*, 2019; Dyer and das Nair, 2013). Health professionals also find it hard to bring up the topic due to personal discomfort (Traumer *et al.*, 2019; Dyer and das

[16] The studies of van den Hondel *et al.* (2015) and Witvliet *et al.* (2018) showed that among female patients, respectively 50%/27.3% (ARM) and 53%/50% (HD) experienced sexual dysfunction and 38%/50% (ARM) and 20%/37.5% (HD) sexual distress. Among the male patients respectively 13.6%/16% (ARM) and 4.4%/11% (HD) experienced erectile dysfunction.

Nair, 2013; Vermeer *et al.*, 2015). Moreover, a lack of knowledge and (conversational) skills (Ho and Fernández, 2006; Hautamäki *et al.*, 2007; Dyer and das Nair, 2013), limited time (Vermeer *et al.*, 2015; van Ek *et al.*, 2015) and resources, giving low priority to the topic, and not feeling responsible have been found to constrain health professionals from providing sexual health support (Saunamäki *et al.*, 2010; Magnan *et al.*, 2005).

To facilitate communication about sexual health during consults various tools have been developed, such as the '(extended) PLISSIT[17] model', 'Talking of Sex' and 'SeCZ TaLK' (Taylor and Davis, 2007; Annon, 1976; Taylor, 2006; Stege *et al.*, 2010; Macdowall *et al.*, 2010). The level of abstractness or practical implementation differ between these models. Although found to be useful for providing care in general, their applicability in the context of ARM and HD is considered limited. Health professionals face difficulties in integrating either of these tools into practice, as also shown by van der Stege *et al.* (2014) in another care context. It is therefore relevant to align the tools to the specific needs of patients with ARM and HD and the health professionals involved. This also requires a better understanding of the mechanisms that underlie the barriers to discussing sexual health (Saunamäki *et al.*, 2010). This study aims to acquire insight into patients' and health professionals' needs to address sexual health in the context of ARM and HD and – from this understanding – to develop a tool that is aligned with patients' and health professionals' needs through a participatory approach.

8.2 | THEORETICAL BACKGROUND

Following the model of Spence Laschinger *et al.* (2010), patients' outcomes are the result of their empowerment, which depends among other factors, on support from health professionals. In turn, for health professionals to feel equipped to support patients, they also need to be empowered. To obtain insights into patients' and health professionals' needs to address sexual health, insights into their empowerment could therefore be helpful.

Health professionals' empowerment is conceptualized at a structural level (the work settings) and a psychological level (health professionals' reaction to these work settings) (Laschinger *et al.*, 2001). To further specify these work settings, the model of Kanter (1987) offers insights. Following this model, four categories of work settings are distinguished: (1) *information*: the access to knowledge about, for instance, organizational policies, goals and values; (2) *support*: the availability of feedback and instructions from managers and colleagues; (3) *resources*: access to requirements, such as materials, money and time needed to fulfil organizational objectives; and (4) *opportunities to learn and grow*: opportunities to learn and grown refer to the availability of rewards and challenges in the work settings. (Kanter, 1987). Using the model of Thomas and Velthouse (1990), psychological empowerment is described as employees' intrinsic motivation in response to the workplace environment, including the following four dimensions: (1) *meaningfulness*: the perceived value

[17] The PLISSIT model has been developed by Jack Annon in 1976 to determine the different levels of intervention in the field of sexuology. The abbreviation stands for Permission, Limited Information, Specific Suggestions and Intensive Therapy.

of certain tasks or goals; (2) *competence*: individuals' belief in their capability to perform their work well; (3) *self-determination*: the degree to which people perceive that they have a choice in introducing and regulating actions; and (4) *impact*: the degree to which people can influence outcomes and issues at work (Thomas and Velthouse, 1990).

To conceptualize the empowerment of patients, the model of Bravo *et al.* (2015) is helpful. According to the model, patients' empowerment level can be determined by two indicators: (1) patients' '*states*' – what patients need to become empowered – and (2) patients' '*behaviours*' – the actions that increase the empowerment 'states'. The 'states' (patients' capacities, states and resources) consist of the elements (1) self-efficacy; (2) knowledge skills, attitudes and self-awareness necessary to influence their own health behaviour; (3) perceived personal control over health and health care; (4) sense of meaning and coherence about their condition; (5) health literacy; and (6) feeling respected. Three types of 'behaviours' are distinguished: (1) shared decision-making; (2) self-management; and (3) patient activation. There is a reciprocal relationship between these indicators. For example, patients' knowledge influences patients' activation, which in turn leads to gaining more knowledge.

8.3 | METHODS

This study was a multidisciplinary collaboration conducted in the period December 2017 – April 2019 in the Netherlands. To guide the study, a project group[18] was set up. The group met before the start of each project phase to discuss the plans and to reflect on the actions and observations of the previous phase.

8.3.1 | PARTICIPATORY ACTION RESEARCH

To develop a tool to empower both patients and health professionals to address sexual health, a Participatory Action Research (PAR) was conducted. PAR is a participatory approach that aims to realize meaningful social change (Shamrova and Cummings, 2017; Baum, 2016; Savin-Baden and Wimpenny, 2007). Because researchers work in close collaboration with relevant stakeholders – in this case patients and health professionals – actions better reflect their knowledge and are better tailored to their wishes and needs (McIntyre, 2008; Baum, 2016). The study consisted of three phases: (1) articulating barriers and (empowerment) needs; (2) translating needs into empowerment tools; and (3) assessing the empowerment tools. In each phase, a cyclical process took place of planning, taking action, observing and reflecting (Gray, 2013). The learnings from each phase were integrated in the subsequent one.

[18] The project group consisted of five patient representatives of three patient organizations, 12 health professionals of six different specialisms of the medical centres Erasmus MC, Radboud UMC and UMC Utrecht, three researchers on patient involvement of Athena Institute and two MSc students of Vrije Universiteit Amsterdam.

8.3.2 | PHASE 1: ARTICULATING EXPERIENCED BARRIERS AND (EMPOWERMENT) NEEDS

Both patients' and health professionals' experiences with sexual health support and their respective needs were identified through an iterative process using a mixed-method design consisting of semi-structured interviews, Online Focus Groups (OFGs) and a questionnaire.

Participant recruitment

Letters were sent to the participants of the study conducted by van den Hondel *et al.* (2015) who indicated that they were willing to participate in further research. Additional letters were sent to individual patients treated at three collaborating medical centres. In addition, patients were recruited via meetings, emails, newsletters and social media of the three patient organizations. Due to a limited response rate, it was decided to conduct mainly interviews (n=11) instead of OFGs. An additional OFG (n=4) was organised to specifically take the perspectives of adolescents into account. These adolescent patients (16–25 years old) were recruited during a weekend trip of two patient organizations. Furthermore, a questionnaire (n=38) was disseminated via the patient organizations to validate the results of the interviews and the OFG. Patients were included who were above the age of 16, treated in the Netherlands and who speak either Dutch or English. In total, 53 patients were involved who live in 11 of the 12 different provinces of the Netherlands. Table 12 gives on overview of the participating patients' characteristics.

Table 12: Characteristics participating patients (n=53)*

Aspect	Category	Interviews	OFG	Questionnaire	Total
Gender	Female	5	3	23	31
	Male	6	1	15	22
Age (years)	16–20	0	3	2	5
	21–30	6	1	16	23
	31–45	2	0	9	11
	45–60	3	0	6	9
	60+	0	0	2	2
	Unknown	0	0	3	3
Diagnosis	ARM	10	2	32	44
	HD	1	2	6	9

Insights into health professionals' experiences and needs were obtained via interviews (n=11) and two OFGs (n=20). All health professionals who are working with the patient groups at the relevant medical centres were contacted via email and/or phone. Recruitment of relevant health professionals working outside the centres was done via snowball sampling. Health professionals of nine different specializations and working at six different medical centres were involved. An overview of the characteristics of all participating health professionals is shown in Table 13.

Table 13: Characteristics participating health professionals (n=31)

Aspect	Category	Interviews	OFG1	OFG2	Total
Profession	Paediatric surgeon	4	0	3	7
	(Paediatric) urologist	1	1	0	2
	Paediatrician	0	1	0	1
	Adult gastrointestinal specialist	1	0	0	1
	Nurse specialist	2	3	4	9
	Gynaecologist	1	1	0	2
	Psychotherapist	1	2	1	4
	Sexologist	1	1	1	3
	Pelvic therapist	0	0	1	1
	Paediatric occupational therapist	0	0	1	1
Deployment	Institutional	11	9	8	28
	Outside the hospital	0	0	3	3

Data collection

The semi-structured interviews took 45–90 minutes. The interviews with patients were structured by discussing their disease and care process by making a timeline. Each step in time was related to their experiences and needs regarding their sexual health.

For the OFGs, a specific tool was used, developed by Nivel (Tates *et al.*, 2009). Because participants are able to join anonymously, this tool has been found useful for discussing taboo subjects, such as sexual health (Tates *et al.*, 2009). In addition, the tool facilitates the participation of patients living in different regions and professionals working at different medical centres. To generate an iterative learning process, the discussions were organised asynchronously. Each OFG lasted for ten days. During this period, participants could log in 24 hours a day. Two researchers moderated the discussions. Regularly new questions were posted. The questions for patients were structured in three parts: (1) experiences with sexual health; (2) experiences with discussing sexual health; and (3) needs to discuss sexual health. To obtain a more in-depth explanation and to stimulate interaction, the moderator posted supplementary questions or asked other participants about their view on a certain subject.

The questionnaire was developed by the online tool Qualtrics and comprised 14 open and closed questions. The questions were based on the outcomes of the previous consultation and were divided into four parts: (1) demographics; (2) experiences with sexual health problems; (3) needs regarding their sexual health; and (4) experiences with and needs regarding sexual health support.

8.3.3 | PHASE 2: TRANSLATING NEEDS INTO EMPOWERMENT TOOLS

Based on the analysis of the findings of phase 1, the project group decided to develop tangible empowering tools in the form of a website for both patients and health professionals. For each tool, a work group was set up consisting of two patient representatives and respectively five and

four health professionals. Among the health professionals, five different disciplines were involved (paediatric surgeon, paediatrician, specialized nurse, sexologist and a psychologist). Two co-creation sessions were organised per work group to translate the needs of phase 1 into the tools which aim to empower both patients and health professionals to address sexual health. The sessions lasted three hours and were moderated by two researchers. During the first sessions, the structure, format and required content of the tangible empowering tools were determined. Subsequently, a draft tool was set up in collaboration with a text writer and a designer. In the second sessions, participants reflected upon the draft and attention was given to language, visualization and implementation. Based on the feedback of the second session and the project group, the tools were improved and integrated into one website.

In addition, the co-creation process was evaluated by an on online questionnaire consisting of five open and four closed questions, of which one included 18 statements. The questions focused on both the process (stakeholders' representation, organization, facilitation and interaction) and the (direct and indirect) outcomes of knowledge co-creation. The questionnaire was completed by ten participants.

8.3.4 | PHASE 3: ASSESSING THE EMPOWERMENT TOOLS

The empowerment tools – in the form of a website – were assessed regarding its user-friendliness and expected (empowerment) outcome. Two online questionnaires were developed via the online tool Qualtrics: one for patients and parents/partners of patients, and one for health professionals. The questionnaires consisted of respectively 27 and 18 closed and open questions. The questions were divided into four parts: (1) demographics; (2) feedback on the website; (3) impact of the website on their empowerment based on the models of Bravo *et al.* (2015), Kanter (1987) and Thomas and Velthouse (1990) for respectively patients and health professionals; and (4) preferences regarding the implementation of the website. The questionnaires were completed by 13 patients, nine parents and 13 health professionals from eight different health care organizations. See Table 14 and Table 15 for respondents' characteristics.

Table 14: Patient and parent respondents' characteristics evaluation questionnaire

Aspect	Category	Patients	Parents	Total
Gender	Female	10	7	17
	Male	3	2	5
Age (years)	0–4	0	0	0
	5–8	0	5	5
	9–12	0	4	4
	12–16	0	0	0
	17–20	1	0	1
	21–30	4	0	4
	31–45	5	0	5
	45–60	0	0	0
	60+	3	0	3
Diagnosis	ARM	11	3	14
	HD	2	6	8

Table 15: Health professional respondents' characteristics evaluation questionnaire

Aspect	Category	Total
Profession	Paediatric surgeon	2
	Paediatric urologist	1
	Gynaecologist	2
	Sexologist	2
	Nurse specialist	1
	Incontinence and stoma consultant[19]	2
	General practitioner	1
Employed at a hospital	Yes	10
	No	3

8.3.5 | DATA ANALYSIS

The interviews were transcribed verbatim. Subsequently, the transcripts of the interviews and OFGs were analysed via the qualitative data-analysis programme MAXqda 2007. A process of inductive and thematic analysis (Green and Thorogood, 2018) was performed by three authors. Deductively, codes were derived from the empowerment models of Bravo *et al.* (2015), Kanter (1987) and Thomas and Velthouse (1990). Inductive coding was used for additional findings. Next, the categorized segments were clustered into sub-themes. Field notes of the co-creation sessions were coded manually based on the coding scheme of the interviews and the online discussions. The data of the questionnaires were analysed via descriptive analysis using Microsoft Excel based on the health professional–patient empowerment models. All categories and themes were discussed among the researchers.

8.3.6 | ETHICAL CONSIDERATIONS

In the assessment of the formal medical ethical committee of Erasmus MC, it was confirmed that the study does not fall within the remit of the Medical Research Involving Human Subjects Act (IRB approval MEC-2017-1133). The study is non-invasive and does not include medical research. The medical ethical committees of two other collaborating centres have followed this advice. The research complied with the national Code of Ethics for Research in the Social and Behavioural Sciences involving Human Participants (VCWE, 2018). All respondents received written and/or verbal information on the voluntariness and incentives of participation, the nature and purpose of the study and the right to withdraw at any time without giving reasons. Verbal informed consent was obtained for recording the interviews and co-creation sessions. All data were anonymized and stored securely. Summaries of the interviews and online discussions were sent to the participants for member check.

[19] In Dutch: WIS (Wond-, Incontinentie- en Stomazorg) consulent

8.4 | RESULTS

The results are described based on the three phases of this PAR study: (1) *articulation of patients' and health professionals' experienced barriers and needs*; (2) *translation of these needs into empowerment tools*; and (3) *assessment of the empowerment tools*. Examples are illustrated by quotes[20].

8.4.1 | PHASE 1: ARTICULATING EXPERIENCED BARRIERS AND (EMPOWERMENT) NEEDS

Patients and health professionals experienced various barriers to and needs regarding patients' sexual health. The barriers and needs are discussed according to the empowerment concept of Bravo *et al.* (2015) for patients, and of Kanter (1987) and Thomas and Velthouse (1990) for health professionals.

Patients' barriers and needs

Patients' barriers and need are presented according to the empowerment 'states' of Bravo *et al.* (2015): *sense of meaning about their condition, feeling respected and knowledge and skills* (see Table 16).

Table 16: Patients' barriers and (empowerment) needs

Barriers	Need	Needs empowerment 'states'*
Avoidance due to distress	Feeling 'normal'	Sense of meaning about their condition
Unaware of possibilities of professional support	Knowledge of possibilities professional support	Knowledge
Difficult to broach the topic	Empathetic response	Feeling respected
Not willing to discuss	Normalizing the (sexual) problems	Sense of meaning about their condition
Unaware of relation between disease and sexual health problems	Knowledge on (possible) sexual health problems	Knowledge
Unaware of possibilities of managing sexual health problems	Knowledge about self-management	Knowledge and skills

Sense of meaning about their condition

More than half of the questionnaire respondents regarded their sexual health as meaningful by indicating its important contribution to their physical and psychological wellbeing. Nevertheless, various interviewed patients addressed that they qualified their sexual health as less important than other needs, such as feeling 'normal' and 'safe', which were seen as more apparent in daily life. The (sexual) problems were seen by patients as not 'normal'. Patients were often not aware that other people (not having the disease) could also experience similar sexual health problems due to other causes. In order to feel 'normal' and 'safe' these patients ignored their (sexual) problems by,

[20] Quotes from the interviews are identified with (I <number>_<role>). The 'number' indicates a specific interviewee and the 'role' whether the interviewee is a patient or a health professional and the health professional's discipline. The quotes from the OFGs are identified with (OFG <number>_RD <number>_<role>), where RD stands for respondent.

for instance, having no sexual contact or not discussing their problems with others. By focusing on other things, patients did not have to deal with their problems. Avoidance was often used as a strategy to cope with their (sexual) problems and reduce stress.

> *It was a kind of mental denial. If I deny it, it will turn out better than expected. If I ignore it, it will prove better than anticipated. That was kind of the line of thought.* (I8_Patient)

In line with this, patient representatives hypothesized that many members of the patient organizations cancel their membership during puberty due to their need to feel 'normal' and not being considered as a 'patient'. In addition, some patients indicated that they saw sexual health as a very intimate or even taboo topic which you do not discuss. Of the questionnaire respondents, less than a half of them (44%) discussed sexual health with their friends, a quarter (26%) with their parents and only a few (17%) with health professionals. A few patients who indicated they had a more open family culture, described their sexual problems as part of their life, sharing experiences with family and friends. Normalizing the topic helped these patients to address them.

> *And I think that if a health professional starts talking about it, it's more likely that you start thinking "Oh it's normal for me to talk." … And if that person starts talking about it, then you think I can also talk about it.* (I6_Patient)

Feeling respected

Another reason for patients to avoid the topic, were feelings of shame and anxiety.

> *Yeah, shame of course. Shame also in general. When discussing the topic with a medical specialist, who is also a person.* (I8_Patient)

Various patients described situations in which they clammed up and therefore were not able to ask questions or join the shared decision-making process. These feelings were reinforced by distrust as a result of bad experiences in the past caused by being 'different'. A few patients who did address their problems during a consultation with a general practitioner or gynaecologist, felt that their concerns were not taken seriously. The health professionals' limited response resulted in patients clamming up, precluding a constructive conversation. Patients addressed their need for a relationship of trust with a health professional with whom they are familiar in order to feel respected and able to discuss the topic.

Knowledge and skills

Patients repeatedly mentioned the need for knowledge and skills. Patients lacked knowledge about the potential sexual problems and the possibilities for self-management and professional support to manage these problems. Fewer than 25% of the questionnaire respondents received information on sexual health, which was mostly regarded as limited and ill-timed. About half of the questionnaire respondents indicated that – due to their lack of knowledge – they were limited in being able to manage their sexual problems. Yet, in time – through their experiences – patients often developed skills and coping strategies to be able to live with their problems. Various patients interviewed, for instance, indicated they obtained control by having sexual contact only when the

problems were limited. They controlled flatulence and faecal incontinence during sex by regulating their diet and managing their toilet visits and bowel enemas. Sexual foreplay, use of lubrication and relaxation exercises helped to relax and prevent vaginism. Most patients avoided one-night stands. Sexual contact was usually only in the context of a long-term relationship, in which trust had been established. Talking with others – mainly their partner helped some of the patients to try to offload their emotional baggage and find a place to put their emotions.

Various patients said that they had (now and/or in the past) limited knowledge of the possibilities of professional support. After the age of 18 years, patients often lacked a contact person at the hospital because the treatment had usually ended. As a result, they did not know whom to contact. The general practitioner was thought to have insufficient expertise. The patients who were still treated at the outpatient clinic were often uncertain whether the first responsible practitioner – due to her or his specialization – was the right person to contact. Paediatric surgeons or urologists were not thought to have knowledge on these topics and patients did not dare to bother them about their concerns. This idea was reinforced by the fact that these health professionals did not mention this topic spontaneously. Patients were often not familiar with the specific expertise of a pelvic physiotherapist or sexologist or found out only later.

> *On my sisters' advice I went to a pelvic physiotherapist. I was 18 years old then. If she would not have told me, I would not have known that such a person existed.* (I2_Patient)

Health professionals' barriers and needs

Only half of the health professionals indicated having discussed sexual health with patients in the past and most of them do not discuss this topic on a regular basis. "*Many topics are tackled, but this [sexual health] is rarely discussed*" (I15_Paediatric surgeon); and "*Structurally discussing [sexual health] is still not common practice*" (FG2_R13_Specialized nurse). Health professionals mentioned various barriers to providing sexual health support to patients. To overcome these barriers, health professionals mentioned various (empowerment) needs at a structural and psychological level (see Table 17).

Structural empowerment

In relation to their structural empowerment, health professionals discussed the need for *information* and *resources*. Regarding *information*, they indicated that clarity is needed about who should be responsible for providing sexual health support. Currently there are different opinions on who should be responsible. Some health professionals thought the paediatric surgeon – as principal provider of health care – should be in charge. Due to the heavy workload of paediatric surgeons and their medical focus, others – including some of the paediatric surgeons – disagreed. Instead, they felt that this was the role of a specialized nurse, who is more accessible and easier for patients to approach. Different opinions were also expressed on whether the responsible health professional should have a long-term relationship of trust with the patient. Some regarded such relationship as beneficial, others as an obstacle for an open conversation on sexual health. A few health professionals mentioned that the whole multidisciplinary team should be responsible. If only one health professional is responsible, it is difficult to guarantee continuity in providing sexual health

Table 17: Health professionals' barriers and (empowerment) needs

Barriers	Need	Empowerment need-level
Responsibilities regarding sexual health unclear	Clear role and task division	Information – Structural
Uncertainties transfers	Information possibilities sexologist / pelvic physiotherapist	Information – Structural
No time to discuss the topic	Time during consults	Resources – Structural
No time to obtain knowledge and skills	Time for professional development	Resources – Structural
Lack of priority given to the topic	Awareness of meaning and importance sexual health	Meaningfulness – Psychological
Difficult to broach the topic	Conversational skills	Competences – Psychological
Difficult to cope with defensive reactions of (parents) patients	Conversational skills	Competences – Psychological
Difficult to diagnose problems	Knowledge about (possible) sexual problems	Competences – Psychological
Difficult to provide support	Knowledge on (possible) solutions for problems	Competences – Psychological
Sexologist involved to limited degree	More central role within patients' care path	Impact – Psychological

support. A need for information was also addressed with regard to whether, when, and to whom patients should be transferred if sexual problems are diagnosed. Health professionals were seldom aware of how a sexologist and pelvic physiotherapist might be deployed. As a result, only a few patients were referred to those specialists.

> *That there is someone like a sexologist is already an eye-opener for some [health professionals]. And what a sexologist does. It would be good to know why you refer someone. … Well, we are not very visible on the digital platform of the hospital.* (I22_Sexologist)

A majority of the health professionals described a lack of *resources* (time and money) to provide sexual health support. According to many health professionals, there is a need for extra time during the consults to discuss sexual health in addition to other topics, such as incontinence, which are seen as having a higher priority. Time is also needed to obtain knowledge on sexual problems and develop conversational skills to discuss sexual health.

> *From the institution, we need an infrastructure to provide education on sexual health to different health professionals of the colorectal team. In addition, at any event one team member should be available for discussing sexuality as common practice during consults, supporting the other team members and to arrange the information provision to patients.* (FG1_R8_Paediatrician)

Psychological empowerment

Regarding their psychological empowerment, the elements *meaningfulness*, *competences* and *impact* were discussed by health professionals. Although most health professionals described sexual health as a *meaningful* topic in (general) patient care, many – apart from nurses – did not regard sexual health as a meaningful topic in relation to their daily practice. Various health professionals mentioned that the topic was not relevant for them because they treated patients before the

age of 12 years. Moreover, sexual health was in general seen by health professionals as having a lower priority than other medical issues such as incontinence. A lack of prioritization was especially apparent for paediatric surgeons and an adult gastroenterologist, who did not regard sexual health as part of their professional responsibilities.

> *I am not a sexologist that deals with the psychological issues. The question is whether I can help them [patients] with their incontinence. When their incontinence improves, they are doing better as well. … We do not treat patients on sexual issues.* (I13_ Adult gastrointestinal specialist)

Several health professionals mentioned that sexual health was not a priority for their patients, because they do not bring it up during the consults. Awareness of the sexual health problems and the need to address them, were mentioned as necessary for prioritizing the topic. Adopting sexual health as a standard topic in patients' medical history and protocols or using a top-three topic list (in which patients can indicate the three main issues they would like to discuss) were suggested as ways to improve prioritization. Yet, some participants indicated that patients will probably feel hesitant to indicate sexual health on the topic list due to feelings of shame.

Various health professionals indicated that they do not feel *competent* to provide sexual health support. When bringing up the topic, health professionals, are confronted with their own interests and values and those of the patient. This is especially the case for male health professionals who have to discuss sexuality with a young girl (especially in relation to the current '#me too' discourse).

> *In daily practice it [sexual health] is not discussed, from both sides. I never had a child that told me that he or she would like to discuss this [their sexual health] with you … that's from both sides. Maybe it is due to my conversational skills. Or the fact that I am dealing with young girls that won't like to discuss such topic with a guy of 40 years old.* (I15_Paediatric surgeon)

Gaining more experience in discussing sexual health – and especially with children and adolescents – would help in dealing with feelings of shame and overcoming discomfort. Health professionals also have to cope with defensive reactions of patients' parents. Preparing parents for a future consultation which will be only with their child due to reasons of privacy, was suggested as a possible solution for coping with parents' resistance. However, not all health professionals agreed on leaving parents outside the consulting room. According to sexologists, parents should be empowered to discuss sexual health with their children by mentioning it from early age. Parents could be informed – via consultations or a group session – about the problems that can arise at the different sexual stages and how they can address these potential problems.

> *I think the subject [sexual health] will be more easily discussed if there is already attention for the topic from early age on. A good moment is when the health professionals informs about primary school. Children with an atypical phenotype or scars get questions from other children: "what do you have there?" It is good that parents are aware that these questions can arise and that it is important for their child to be able to answer those questions.* (FG1_R9_Psychologist)

In addition, most of the health professionals indicated that they did not know what would be a

good 'age' to address sexual health. Some thought just before puberty, others at the age of 12, after achieving continence – because of the priority given to continence before that time – or at the age of three, because of the start of children's sexual development. A few health professionals mentioned that, in order to be empowering, the moment of introducing the topic should depend on the child's development. According to sexologists, knowledge about the stages of sexual development would be helpful in improving the timing of the support provided by health professionals. This knowledge was also found to be worthwhile for identifying problems (at an earlier stage). The development of a negative body image during childhood can, for instance, hinder starting intimate relations during adulthood. Underdiagnoses of sexual health problems by health professionals was also thought to be caused by framing sexuality only in relation to sexual intercourse.

> *Many health professionals stop after questioning whether you are sexually active. That [if you are not sexually active] does not mean that you do not have any sexual feelings or thoughts. It does not cross their mind that you can be sexually active in your mind and that you can be a sexual person. Everyone experiences sexuality differently. Irrespective whether you are sexually active.* (I12_Paediatric urologist)

Moreover, health professionals described a need to have greater knowledge about sexual health problems in patients with ARM and HD and possible solutions through, for instance, training or information to be able to diagnose sexual health problems and provide support.

8.4.2 | PHASE 2: TRANSLATING NEEDS INTO AN EMPOWERMENT TOOLS

The results of the first phase show patients' need for normalizing the topic, feeling respected and having knowledge and skills; and the need of health professionals to have clear arrangements about the possibilities, knowledge, skills, resources (time) and impact (in the case of sexologists) on how sexual support is provided. Based on these empowerment needs, we decided to develop tangible empowerment tools for patients, their parents and health professionals. The empowerment tool for patients and patients' parents aimed to educate patients and their parents and stimulate individualized verbal support by increasing patients' help-seeking behaviour. Because of patients' preference for digital information – for privacy and easy availability – an online tool was created. In addition, online information was developed for health professionals to obtain more knowledge and competences to discuss the topic, identify problems and to empower patients by providing support. This information should also provide tools for health care organizations to facilitate the integration of sexual health issues within current protocols.

Co-creation sessions were organised to translate the needs from phase 1 into these empowerment tools by discussing their format and content. The tools 'Talking of Sex' and 'SeCZ talk' were respectively not available for the Dutch context and not applicable for individual patient consultation. Patients furthermore indicated that due to feelings of shame, group meetings were not their preference. The PLISSIT model was suggested as a useful guideline for discussing sexual health, but the participants found it very generic and lacked specific tips for patients with ARM and HD.

Based on patients' needs, the website for patients should aim to: (1) increase knowledge on sexual

health and opportunities for self-management; (2) share experiences; (3) normalize the problems; and (4) provide support consisting of tips and suggestions for contacts. The normal psychosexual development stages were found to be an appropriate basis for structuring the website. A neutral though positive tone was chosen for the texts to prevent being perceived as over-dramatic. Based on health professionals' needs, the digital information for health professionals aimed to: (1) increase awareness about the importance of patients' sexual health among health professionals; (2) normalize the topic; and (3) provide tools to facilitate discussion of the topic, to identify problems and to support patients and parents. The information was structured based on the organizational and professional barriers to integrating sexual health as a standard of care. To make the information more accessible outside the hospital setting, we decided – based on discussions with the project team - to integrate both tools into a digital information source.

Participants provided different types of input, which complemented each other. Patients mainly addressed the type of sexual problems experienced and the importance of personalization and transmural support. Health professionals' input was mostly related to the feasibility and practicalities regarding implementation. In developing the materials, a balance between these different types of input was found. For instance, the use of the term 'vulva' or 'vagina' was discussed. Vulva was the correct term, but vagina was commonly used by most health professionals and familiar to patients. It was decided to use the term 'vagina' in line with other related materials. Another example was the solution to advise health professionals to start the conversation at an early age so that patients and their parents become familiar with the topic, for the discussion whether to advise health professionals to exclude parents when the topic is introduced.

Participants found the sessions valuable. All participants felt involved and taken seriously n=10 (100%), and almost all n=8 (80%) thought that the sessions led to more insights into the different perspectives and the integration of these perspectives into a better result.

> *Very meaningful. The interaction of health professionals from different disciplines and experience experts led to an efficient exchange of ideas and perspectives. As a result, decisions on the use of materials for the website could be easily made* (Paediatrician).

Based on the input of phase 1 and 2, a draft website was developed by a designer and a text writer in collaboration with the researchers. The website consisted of six sub-parts: (1) general information, and information for (2) parents, (3) children, (4) adolescents, (5) adults, and (6) health care providers. It was decided to not to add ages to categories 3, 4 and 5, taking into consideration that someone's sexual development can develop differently and giving patients the possibility to decide for themselves which phase is most appropriate to their situation. The general information consisted of an overview of possible sexual problems, relevant health providers, other relevant websites, and a glossary. Gender-specific information was provided for both patients and their parents. Parents' information was based on the stages of normal sexual development and divided into three parts: (1) 'normal' sexual development; (2) possible (future) problems related to the disease; and (3) tools to empower their child in coping with these problems. The information for adolescents and adults was structured based on the questions they addressed. Specific information was provided for health

professionals to increase awareness and improve their knowledge and conversational skills (based on their needs for psychological empowerment). In addition, information was provided for health providers focused on the aspects 'information' and 'resources' of structural empowerment. The final Dutch version of the website is published at www.seksualiteit-arm-zvh.nl.

8.4.3 | PHASE 3: ASSESSING THE EMPOWERMENT TOOLS

The draft website was tested for its expected empowering effect on both patients and health professionals.

Patients' empowerment

In general, patients and patients' parents positively evaluated the website (intelligibility n=21/22 (96%), accuracy n=20/22 (91%) and completeness n=18/22 (82%)). Regarding their empowerment, the patients anticipated that the website will have a positive effect on their knowledge (67%)[21], on how they will regard sexual health (62%) and whether they feel respected by health professionals (56%). A slight positive effect was expected regarding health literacy (44%) and perceived positive control (44%).

> *I am very happy that there is – at last – attention for it [sexual health] and I got more clarity on the relation to Hirschsprung Disease. Now I understand why I had difficulties with being touched during relationships. It is rather emotional to find out the underlying reason of this problem.* (Patient)

Patients' parents expected that the website would positively affect their empowerment through greater knowledge (89%)[21], self-efficacy (78%), meaningfulness (78%), health literacy (67%) and feeling respected (63%) and perceived personal control (59%).

Health professionals' empowerment

Although in general positive about the website – intelligibility n=12/13 (92%), accuracy n=11/13 (85%) and professional n=11/13 (85%) – health professionals addressed various points of improvement regarding, for instance, reducing the amount of text. Regarding their empowerment, health professionals often indicated 'neutral' or 'not applicable', especially with regard to their resources (67%)5, support (62%) and competences (62%). The number of neutral answers was higher among health professionals working outside the hospital because the work settings (structural empowerment) are probably less applicable to them. In addition, the need for competences is not relevant for sexologists, given that they are specialized in the topic. Diversity in responses were found with regard to information (39% agree and 18% disagree) and self-determination (26% agree and 23% disagree). This diversity is probably due to differences in personal and professional characteristics, such as job function. An expected result of the website on health professionals'

[21] The percentages of the website its' expected effect on the empowerment of patients, patients' parents and health professionals, are each based on three questions. In total 13 patients, 9 parents and 13 health professionals filled in the questions.

empowerment was found regarding their meaningfulness (67%), opportunities to learn and grow (62%) and possibly their expected impact on providing sexual health support (46%). Yet, as one of the respondents mentioned, '*A website cannot solve all the problems*'.

8.5 | DISCUSSION

In this study we aimed to develop a tool that is aligned with patients' and health professionals' needs to address sexual health in the context of ARM and HD. We showed that a holistic approach is needed, which integrates the needs of patients, their parents and health professionals to address sexual health issues. The use of empowerment models to guide the assessment of patients' and health professionals' needs enabled them to be translated into an empowerment tool. In line with the need for more education on sexual health highlighted in previous studies (Saunamäki *et al.*, 2010; Carlsson Wincrantz and Wahlberg, 2011; Mellor *et al.*, 2013), the empowerment tool – developed in this study – provides information for patients, their parents and health professionals. In comparison with existing tools, this tool – in the form of a website – adds the integration of patients', parents' and health professionals' needs in a co-creation process. By specifically focusing the website on specific disease types and cultural context, the tool is better aligned with patients' and health professionals' needs (Goossens *et al.*, 2011). In contrast to the PLISSIT model – which focuses on one-way communication (Mercer, 2008) – the tool facilitates a collaborative dialogue. The use of the empowerment models enabled the assessment of the tool's expected impact on both patients', patients' parents' and health professionals' empowerment and assisted in bringing about a change needed to implement the tool in practice. The implementation of the tool is facilitated by aligning it to the specific context of ARM and HD care and generating national support through the participatory approach of this study. In addition, the outcomes of this study will be included in the Dutch standard of care of ARM and HD. Addressing the topic in the clinical guidelines will help in anchoring the tool in practice, which has been identified as a barrier for existing tools (van der Stege *et al.*, 2014).

To address their sexual problems, patients indicated a need to feel 'normal', have an empathetic response and have knowledge about potential sexual problems and possibilities for self-management and professional support. A knowledge gap among patients with ARM and HD is in line with the findings of Hartman *et al.* (2006), who found a lack of general information provision among the patient groups. Patients' stories also resonate with the hypothesis of Hartman *et al.* (2008) that 'adolescents have a strong wish to be 'normal', possibly explaining that patients' filled in the questionnaire in a socially desirable way (Hartman *et al.*, 2008). This study furthermore adds patients' need to become more knowledgeable about how to seek help and to be comfortable with and able to discuss their sexual health. Parents could help their children by supporting an open conversation on sexual health from an early age.

To empower patients to obtain support, health professionals should be empowered at a psychological level by improving the way they prioritize sexual health, increasing their knowledge, conversational skills and sexologists' 'visibility'. At a structural level, health professionals should be empowered by arranging responsibilities regarding sexual health, information on when to transfer a patient to a

sexologist and time to obtain education and implement sexual health as a topic during a consult. Lack of competences, time and resources and feeling that their responsibility was limited were also highlighted in previous studies as barriers to health professionals provision of sexual health support (Hautamäki *et al.*, 2007; Stead *et al.*, 2003; Ho and Fernández, 2006; Dyer and das Nair, 2013; Vermeer *et al.*, 2015). This study specifically addresses the 'invisibility' of sexologists in the hospital. Health professionals have been found to have limited knowledge about how to deploy sexologists. In addition, sexologists were seen to have limited impact on putting sexual health on the agenda. This resonates with the study of Alarcão *et al.* (2016: p.1190) who showed that sexology is not seen as a "*full-fledged profession but rather as a specialization or a secondary field of action*" due to the historical development of the specialization from different backgrounds – psychologists, physicians or nurses (Fugl-Meyer and Giami, 2006).

The website is expected to increase awareness, fill current knowledge gaps among patients, patients' parents and health professionals and to improve their skills. It is expected that this knowledge will broaden stakeholders' views on sexual health (being more than sexual intercourse) (Hordern and Street, 2007), the importance of sexual health for patients' well-being and normalize the inclusion of topic in patient–health professional interactions (Mellor *et al.*, 2013), which is necessary to overcome restrictive attitudes to sexual health in the context of chronically ill patients (McInnes, 2003). Resources (time and money) are also needed so that health professionals can receive sufficient training (Jaarsma, 2011) to provide sexual health support or to integrate such support in current protocols. Previous studies indicate that structural empowerment underlies health professionals' psychological empowerment (Knol and Van Linge, 2009; Wagner *et al.*, 2010).

Strengths and limitations

A strength of this study is the nationwide collaboration of different academic medical centres and patient and professional organizations to connect insights into patients' and health professionals' empowerment needs, enabling the development of suitable empowerment strategies. A large proportion of the health professionals working with the patient groups in the Netherlands were involved. The relatively high response rate (60%) of health professionals indicates their interest in the topic. By using a variety of research methods, the lack of support regarding sexual health among the patient groups – showed by van den Hondel *et al.* (2015) – was further explored. In addition, by co-creating the materials, a widely supported website could be developed which contributes to the successful implementation of the tool. The website was translated into English to make it accessible for non-Dutch-speaking patients.

A limitation of the study is that for Hirschsprung Disease, only nine patients participated in the first phase of this study. The difficulty in recruiting patients for the OFG and interviews might be explained by the taboo nature of the subject. Parents are important stakeholders in the empowerment of patients during childhood. Efforts were undertaken to recruit parents at the data collection in phase 1, yet no-one applied. Patients' parents' perspectives were, however, taken into account in phase 2 and 3 and throughout the project via the project group. Furthermore, the experiences and needs of non-Dutch or English-speaking and/or digital-illiterate patients were not taken into account. The tool assumes patients have ICT resources and skills, thereby excluding patients with a lower income,

education or literacy and further exacerbating their vulnerability.

8.6 | CONCLUSION

Patients and health professionals were found to have limited knowledge and skills to address the sexual health of ARM and HD patients. In addition, patients' and health professionals' impeding attitudes towards sexual health, health professionals' lack of time and sexologists' limited involvement in the care pathway, hinder their empowerment. By simultaneously educating patients, patients' parents and health professionals, the empowerment tool – in the form of a website – is expected to change current attitudes to addressing sexual health. Subsequently, the tool is expected to help in legitimizing the topic, which is necessary for the allocation of resources and increasing the power of sexologists within current care pathways. This tool could be used as a format for other disease types.